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Hydatidiform Mole Mimicking an Enlarged Uterine Fibromyoma Four Months After ART

Case Report

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Abstract: Hydatidiform mole is a pregnancy disorder, of a benign nature. We present a case of molar tissue within a uterine myoma, the first such entity reported in the literature. In May 2006, a thirty-eight year old infertility patient was admitted for myomectomy. She had anamnesis for chronic pelvic inflammatory disease and surgeries performed for tubal pregnancies. After the sixth intracytoplasmal spermatozoa injection procedure performed in January 2006, she conceived, but curettage was performed in March 2006 for a missed abortion. Following the routine preoperative evaluation in May 2006, four months after the last artificial reproductive technology procedure, myomectomy was performed as uneventful operation, but the histological report appeared unusual, showing degenerated chorionic villi within the uterine myoma. Molar tissue within uterine myoma might evolve even after artificial reproductive procedures. Furthermore, this finding might be misinterpreted as a fibromyoma degeneration. This is the first, and a unique case, of molar tissue within uterine myoma reported in the literature.

Keywords: *Hydatiform mole • Infertility • Uterine myoma • ART • Surgery*

1. Introduction

Hydatidiform mole presents as an abnormal pregnancy characterized by degeneration of the chorionic villi [1]. The disorder is usually of benign nature and remits after evacuation of the uterine cavity [2]. Rarely, however, it can also penetrate the uterine wall and spread into surrounding tissue, or get into the veins and, through blood flow, migrate to other distant organs [3-5]. We present a case of molar tissue within a uterine myoma, which is, according to the data available at present, the first such entity in the literature.

2. Case Outline

An infertility patient, 38 years old, was admitted for surgery for uterine myoma. Apart from infertility that had lasted for 15 years, her personal/family history was found to be insignificant for the actual disorder, while data regarding her bleeding pattern were typical for oligomenorrhoea.

The patient had a history of 4 laparotomies and 2 laparoscopic surgeries to treat different problems. In 1992, her right Fallopian tube was partially removed due to an ampullary (ectopic) pregnancy. Only twelve months later, in 1993, the remaining part of the right tube had to be removed because of the repeated ectopic pregnancy. As she had been suffering from infertility throughout the whole period and all known causes had been excluded, in 2002 she had laparoscopy followed by a laparotomy. During that procedure, the

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left tube was found to have developed a hydrosalpinx, and salpingoophorolysis and salpingoneostomy were performed; these were typical procedures for that period and were performed according to the patient's written wishes. During preoperative evaluation for this procedure, an asymptomatic intramural formation 18 mm in diameter having sonographic features typical of myoma, was found in the uterine fundus, without accompanying secondary changes.

The second laparoscopy, performed in October 2005, revealed a repeated hydrosalpinx of the left tube. The procedure was converted to the fourth laparotomy, adhesiolysis, and salpingectomy.

Only one month after that surgery, the patient underwent the long protocol of IVF-ET (in-vitro fertilization and embryo transfer) procedure, and conceived after the sixth cycle in January 2006. Human chorionic gonadotropin (HCG) levels were within normal ranges for the gestational week. However, repeated ultrasound evaluation in the early pregnancy confirmed a missed abortion. Therefore, evacuation of the uterine cavity by curettage was performed in March 2006, and histological examination of the specimen obtained revealed findings typical of the diagnosis.

After this procedure of pregnancy termination she underwent regular checkups. Significant enlargement of the uterine myoma, from 18 mm up to 55 mm, was sonographically confirmed in April 2006, with secondary changes typical of degeneration. Therefore, in May 2006, four months after the last ART (artificial reproductive technology) procedure, and two months after pregnancy termination by curettage, our patient was admitted for surgery. During preoperative evaluation all necessary analyses were done, and found to be within normal ranges, except for fibrinogen and C-reactive protein, which were 8.94 g/l and 89.4 mg/l, respectively.

The fifth laparotomy was performed and a myoma located in the uterine fundus, in the posterior wall and to the right corneal region, 55x50x45 mm in diameter, with macroscopic degenerative changes, was removed. Apart from myomectomy, adhaesiolysis/oophorolysis was also performed. Both surgery and recovery were uneventful.

The removed uterine myoma was analyzed histologically. The histological report was surprising, describing degenerated chorionic villi within the tissue of the uterine myoma. Histopathological findings included an ultrastructure of complete molar cystic villi with tree-like branching of the microvillus processes and intracytoplasmic lacunae without capillaries in the stroma. Large lacunae with microvilli and polymorphic nuclei of syncytiotrophoblast cells with well-developed organelles were also observed.

Her HCG level was regularly followed-up after surgery, from the period the diagnosis was established up to the 6th month after the procedure. The initial HCG concentration two weeks after the last surgery, when the diagnosis was established, was 450mIU/ml, and showed a steady-state decrease during two-week periods; levels were within the normal range as early as 2 months after myomectomy. The HCG concentrations remained within normal range throughout the next 4 months. Furthermore, our patient's HCG levels were checked at six-month intervals along with a sonographic scan of her uterus, and found to be completely normal. Nowadays, more than 4 years after the last surgery, she has requested the next ICSI procedure.

3. Discussion

According to the recent literature data, hydatidiform mole shows incidence rates of 220/100,000 pregnancies, 264/100,000 total births, and 266/100,000 live births. However, some authors have published findings of rates of hydatidiform mole incidence that are even higher [6]. Hydatidiform mole usually occurs in multiparous women under 20 or over 35 years, of Caucasian or Asian origin, and especially in those with history of previous molar pregnancy [4,6]. As maternal age is confirmed to be a significant risk factor for molar pregnancies [6], our patient also belongs to this group, being Caucasian and 38 years old.

There are several problematic points at which a diagnostic error might occur: e.g., the diagnosis of early complete mole as partial mole; the over-diagnosis of hydatidiform mole in tubal pregnancy; and the diagnosis of placental site non-villous trophoblasts as a placental site trophoblastic tumor or choriocarcinoma, particularly if associated with atypia, as frequently observed in a complete mole [6]. Nowadays, ultrasound scan has proved a promising diagnostic tool in almost all patients with a suspected invasive mole [5]. However, in very rare cases, the correct diagnosis is missed, and the scan is wrongly interpreted [4,5]. Such circumstances happened in our patient where molar tissue within uterine myoma was first described as degeneration of a fibromyoma. Therefore, histological investigation of the tissue removed still remains the final diagnostic tool [7].

There have been cases of molar tissue described in the cornual regions of uterus [8,9] or those that have spread to the cervix and tubes [9]. In some rare cases the molar tissue can be found in organs surrounding the uterus (vulva, vagina, Douglas pouch, broad ligament) or distant structures (lungs, brain and even surgical scars) [2,4].

Hydatidiform mole has also been reported in ectopic pregnancy. In one large retrospective study carried out on 132 patients with ectopic tubal molar pregnancy, all cases were managed surgically, without development of persistent gestational trophoblastic disease and with the HCG concentration spontaneously returning to normal values [10]. This post-interventional HCG follow-up after molar pregnancy is recommended to rule out the development of post molar gestational trophoblastic neoplasia [1]. Accordingly, in our patient, postoperative serial serum HCG concentrations were regularly checked; they showed a steady decline from levels of 450mIU/ml to normal values in a period of two months, and remained within normal range throughout the next 4 months, and in all further six-month check-ups, until the end of 2008. It seems that the immunohistochemistry of molar degeneration of trophoblastic tissue would be more useful for a final diagnosis [11].

Molar tissue is also being found in twin pregnancies that were conceived naturally or by an ART technique, where a complete hydatidiform mole coexists with a viable twin. Although most of these pregnancies are prematurely terminated, there still exists a chance to deliver a healthy infant [12-14]. It is believed that twins in such cases most often derive from one single oocyte fertilized by one spermatozoon [14].

Intramural molar pregnancy has also been reported in patients with an incomplete abortion [2], and in pregnancies achieved either by natural conception or by an ART procedure [14]. Intramural molar pregnancy was described in a 28-year-old woman in her fourth pregnancy after evacuation of retained products of conception for an incomplete miscarriage at 8 weeks' gestation that occurred 2 months earlier. In this patient, an ultrasound scan demonstrated a complex mass within normal myometrial tissue of the uterine fundus [3]. Contrary to Bhatia's report, although in our patient the diagnosis was also sonographically suspected two months after the evacuation of the products of conception from the uterine cavity, the disease appeared after missed abortion and within a uterine myoma [3,5].

The case we report here also demonstrates that even despite apparently normal embryos being implanted during an IVF/ICSI/ET procedure, molar pregnancies might still appear. Nevertheless, it is highly unlikely that this complication is related to the technique of IVF/ICSI/ET itself, but rather is the result of the characteristics of the women undergoing ART procedure and their partners that make them more at risk for molar pregnancies [14,15].

In conclusion, molar tissue within uterine myoma might occur, even after an IVF/ICSI/ET procedure. Furthermore, this finding might be misinterpreted as fibromyoma degeneration. According to the data available, this is the first and a unique case in the literature of molar tissue within a uterine myoma.

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